CASE REPORT

Recurrence of Graves' disease in ectopic thyroid tissue

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Accepted 8 January 2018

SUMMARY

A 40-year-old woman with a history of Graves' disease status postorbital decompression for severe ophthalmopathy underwent total thyroidectomy by a high volume thyroid surgeon in July 2013 with a benign final pathology. Eight months later, she presented with a mass on the right anterior neck that showed minimal growth over time. Her thyroid stimulating immunoglobulin and thyroid-stimulating hormone receptor antibody levels were consistently elevated and increasing. She underwent removal of the neck mass in September 2016. Final pathology showed benign thyroid tissue with diffuse hyperplasia and lymphoid follicles, consistent with Graves' disease. We present an unusual recurrence of Graves' disease post-total thyroidectomy that recurred secondary to ectopic thyroid tissue in the right upper anterior neck deep to the strap muscles.

BACKGROUND

In treating Graves' disease, total thyroidectomy appears to offer the least recurrence rates among treatment options. Despite this, there have been cases of Graves' disease after total thyroidectomy. Rates of recurrence are variable from 0% to 33%.^{1,2} Graves' disease after total thyroidectomy (removal of all visible thyroid tissue in the neck) can recur in the thyroid bed if tissue is missed during surgery or in ectopic thyroid tissue. Ectopic thyroid tissue is a result of abnormal thyroid gland descent during

Figure 2 Ectopic thyroid mass (arrow) in sagittal plane.

gestation and usually appears at the midline or just lateral to the midline along the thyroglossal tract. The exact incidence of ectopic thyroid tissue is not known, likely because it is often asymptomatic. Ectopic thyroid tissue can be confused with thyroglossal duct cysts, which are expansions of the thyroglossal tract that can sometimes be lined with thyroid tissue. On our literature review through PubMed, there were no reported cases of Graves' disease from thyroid tissue in the lining of a thyroglossal duct cyst, although there was a report of hypothyroidism postexcision of thyroglossal duct cyst.³

CASE PRESENTATION

A 40-year-old woman with no significant other medical conditions or social history and a family history consisting of hypertension, sickle cell disease and some unknown type of thyroid problem in an aunt, presented in 2011 with symptoms of palpitations, periorbital oedema, painless goitre, tremor and weight loss. Her lab tests, in January 2012, showed hyperthyroidism with a thyroid-stimulating hormone (TSH) level of <0.008 uIU/mL (normal 0.20–4.78 uIU/mL), a total triiodothyronine >800 ng/dL (normal 60–180 ng/dL) and a free thyroxine of 6 ng/dL (normal 0.8–1.8 ng/dL). Thyroid ultrasound demonstrated a diffusely enlarged, heterogeneous thyroid gland with multiple irregular hypoechoic areas scattered throughout the thyroid gland and colour Doppler showed increased vascularity. Thyroid scintiscan showed diffusely increased uptake of iodine in the thyroid gland (figure 1). All of these findings were consistent with Graves' disease. She was started on methimazole 10 mg three times a day as well as propranolol 40 mg four times a day and showed improvement in symptoms.

In February 2012, she underwent bilateral frontal sinusotomy, bilateral total ethmoidectomy, bilateral sphenoidotomy and bilateral maxillary antrostomy with medial and inferior floor decompression of the orbit after receiving intravenous

Figure 1 Initial thyroid uptake scan showing Graves' disease in 2011.

To cite: Jakibchuk K, Ali S, Samantray J. *BMJ Case Rep* Published Online First: [please include Day Month Year]. doi:10.1136/bcr-2017-221566

Figure 3 Ultrasound of ectopic thyroid tissue with colour and power Doppler.

methylprednisolone. Her CT and MRI of the orbits, without contrast, had shown bilateral compression of the optic nerve at the orbital apices secondary to diffuse extraocular muscle enlargement and mild orbital fat stranding. Eight months later, she required further bilateral orbital floor and lateral wall decompression as CT of the orbits showed interval progression of the thyroid orbitopathy.

In July 2013, she was hospitalised with compressive symptoms in her neck and an elevated free thyroxine of 2 ng/dL with a TSH level of 0.011 uIU/mL. She underwent total thyroidectomy that admission, with final pathology showing diffuse hyperplasia consistent with Grave's disease. Post-thyroidectomy TSH was 12.098 uIU/mL and total thyroxine 3.7 mcg/dL (normal 6.2–14.6 mcg/dL). In March 2014, she felt a lump on the right upper anterior part of her neck despite having undergone total thyroidectomy. She denied dysphagia, dysphonia, hyperthyroidism symptoms or changes in vision. Furthermore, she was requiring decreasing doses of levothyroxine until it was discontinued.

INVESTIGATIONS

Ultrasound imaging in October 2014 showed a 2.0×1.7×0.8 cm ovoid isoechoic heterogeneous solid mass in the right anterior neck, deep to the strap muscles (figure 2), which had exuberant vascularity on colour and power Doppler (figure 3). In November 2015, she started reporting some dysphagia to solids and liquids. Neck ultrasound in July 2016 indicated that the mass had minimally increased in size from 2 cm to 2.2 cm. Her thyroid stimulating immunoglobulin (TSI) and thyroid-stimulating hormone receptor antibody (TRAb) levels were consistently elevated and increasing. At that time TSH receptor antibody levels were 4.33 IU/L (normal <1.75 IU/L) and TSI was 399% (normal <123%).

TREATMENT

She underwent removal of the ectopic thyroid tissue on the inferior constrictor muscle of the right side of her neck in September 2016. The mass was solid and final pathology showed benign thyroid tissue with diffuse hyperplasia and lymphoid follicles, consistent with Graves' disease.

OUTCOME AND FOLLOW-UP

Postoperatively, her parathyroid hormone was 74 pg/mL (normal 18–86 pg/mL). The bilateral vocal cords were noted to be mobile and she was discharged home after an uneventful postoperative course on 75 mcg of levothyroxine daily. In November 2016, labs were significant for a TSH level of 6.518 uIU/mL and a free thyroxine level of 1.3 ng/dL.

DISCUSSION

In reviewing the literature, Graves' disease recurrence post-total thyroidectomy from hyperfunctioning ectopic thyroid tissue is

Learning points

- ▶ There are few reported cases of Graves' disease in ectopic thyroid tissue post-total thyroidectomy.
- ▶ If a patient is on thyroid hormone replacement and becomes hyperthyroid post-total thyroidectomy, medications should be slowly weaned to rule out iatrogenic causes, as were in this case.
- ▶ Remnant or ectopic thyroid tissue after total thyroidectomy may become hyperfunctional in the presence of stimulating thyroid-stimulating hormone receptor antibody and grow in size, presenting as a mass.
- ▶ In patients with symptoms and lab tests indicating hyperthyroidism post-total thyroidectomy, imaging will help diagnose Graves' disease recurrence in ectopic thyroid tissue.

rare. There are a few individual case reports of Graves' disease recurrence post-total thyroidectomy from a hyperfunctioning midline neck mass, as small as 4 cm, described as a thyroglossal duct remnant.^{4–7} One case described a mediastinal mass with functioning thyroid tissue.⁸ These masses were evaluated with technetium or radioactive iodine uptake scans and/or TRAb levels. There was one particularly unique case of Graves' disease in ectopic thyroid tissue reported after prior removal of ectopic tissue at the same site 9 years previously.⁹ The patient had no previous history of Graves' disease.

One retrospective case series looked at 3044 total or secondary total thyroidectomies performed for benign multinodular goitre and found that only 1 out of the 10 recurrences was in the thyroid bed.¹⁰ Four were in the pyramidal tract and five in the thyrothymic tract. They concluded that a clear understanding of embryology is essential to successful total thyroidectomy and that this understanding would reduce recurrence rates. This is important because it will prevent the need for repeat surgeries that can lead to greater healthcare costs and greater risk for patients with Graves' disease.

Acknowledgements This case report was reproduced from Kalyna Jakibchuk *et al.* SAT 188: Recurrence of Graves' disease in ectopic thyroid tissue. *Endocrine Reviews* (2017).

Contributors Substantial contributions were made by all authors in the creation of the manuscript. All authors contributed in editing the submitted article, approved the final version and agreed to be accountable for the work.

Funding This research received no specific grant from any funding agency in the public, commercial or not-for-profit sectors.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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